DISCUSSION

There is an element of uncertainty in suggesting that the pulmonary granulomatosis (thesaurosis) in this case was produced by the excessive use of body sprays. It is regrettable that qualitative tests for polyvinylpyrrolidone were not performed on the lung tissue removed for biopsy. Bergmann[2] said he considers this test necessary for an absolutely certain diagnosis, as he and his coworkers have apparently incriminated this ingredient by chemical analysis of various sprays as responsible for the changes evoked by prolonged inhalation.

The pathologic changes suggesting the diagnosis are shown in Figures 5 and 6. The lesion is practically indistinguishable microscopically from sarcoidosis except for the presence of the PAS-positive granules in the histiocytes. These granules were seen in all the cases Bergmann[2] observed. It is significant that we were unable to demonstrate similar PAS-positive granules in specimens from known cases of sarcoidosis selected from our pathologic files. In view of these observations, and in the absence of other possible etiologic agents, the history of excessive exposure to spray type cosmetics certainly seems more than coincidental.

It is noteworthy that in the present case more than a year elapsed before the pulmonary lesions cleared, whereas it took an average of only three months after discontinuance of sprays in the two cases originally reported by Bergmann.[1] Our impression following correspondence with Bergmann, however, is that the lung reaction in the present case was pathologically and radiographically more extensive and intensive than in most of the cases he observed.

The virtual complete resolution of the lesions on the radiograms is by no means specific for thesaurosis. However, this degree of resolution without steroid therapy would be regarded as most unusual in extensive pulmonary sarcoidosis, which in our opinion is now the only significant entity to be considered in the differential diagnosis.

Obviously more data must be accumulated regarding this probable entity before final conclusions can be drawn.

SUMMARY

In a case of diffuse bilateral pulmonary granulomatosis, the lesions disappeared without therapy approximately 14 months after the patient stopped using cosmetic body spray that she had previously used often. Scalene node biopsy and lung biopsy revealed a granulomatous reaction identical with that previously described following the use of hair spray. This factor should be considered in patients with asymptomatic pulmonary disease.

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REFERENCES

1. Bergmann, M.: Flance, I. J., and Blumenthal, H. T.: Thesaurosis following inhalation of hair spray, N.E.J.M., 258-471-476, 1958.

2. Bergmann, M.: Personal communication.

Hemolytic Disease of the Newborn Due to Sensitization to the Blood Factor hr'

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SINCE THE DISCOVERY of the hr' factor by Levine in 1941,[5] approximately 30 articles concerning hemolytic disease of the newborn due to sensitization to this factor have appeared in the literature.

In view of the relative infrequency of hemolytic disease of the newborn related to maternal sensitization to the hr' factor it seems important to report the following case.

REPORT OF A CASE

The patient, obese, 37 years of age, gravida IX, para V, abortus III, was admitted to Kern County (California) General Hospital on June 1, 1960. The expected date of confinement was June 17, 1960. None of her five living children, whose birth weights had ranged from 7 pounds to 9 pounds 8 ounces, had jaundice in the newborn period and all were well. In none of the three cases in which abortion occurred did gestation continue more than a few weeks and the cause of abortion was not known.

The patient had received three blood transfusions —January 25, 1950, after an abortion; September 27, 1957, and September 28, 1957, for postpartum hemorrhage. Since there was a family history of diabetes mellitus a glucose tolerance test was done May 27, 1960. The results were indicative of latent diabetes mellitus. A 1200-calorie diet was prescribed and the patient was observed regularly in the Diabetic Clinic. Blood sugar content remained within normal limits during that time.

On June 3 the patient was delivered of a 9-pound 3-ounce edematous girl with grayish-blue discoloration of the body that was attributed to a somewhat difficult delivery. Cyanosis of the face was also noted. Studies of the infant's blood done the next morning showed a positive reaction to a Coombs test; serum bilirubin of 23.7 mg. per 100 cc.; hemoglobin, 15.3 gm. per 100 cc.; hematocrit, 53 per cent; 3 nucleated red blood cells per 100 white blood cells. The blood was typed as Group O, Rh_0-positive, hr'-positive. The mother's blood was Group B, Rh_0-positive, hr'-negative. It was believed that the infant had hemolytic disease of the newborn due to maternal sensitization to the hr' factor. An exchange transfusion was done, using 500 cc. of Group O, Rh_0-positive, hr'-negative blood and the infant tolerated the procedure well. The bilirubin was 24.6 mg. per 100 cc. before the exchange and 13.6 mg. after it. On the morning of June 5, 1960 the bilirubin was 22.4 mg. per 100 cc. A second exchange transfusion was done, using 500 cc. of Group O, Rh_0-positive, hr'-negative blood, without incidence. The bilirubin content was 25.6 mg. per 100 cc. before and 14.6 mg. after the exchange. On the morning of June 6, 1960, the bilirubin was 21.4 mg. per 100 cc., the

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hemoglobin concentration was 12.8 gm. per 100 cc. and the hematocrit 41 per cent. At 2:30 p.m. the bilirubin was 21.5 mg. and at 8:30 p.m. it was 20 mg. per 100 cc. At 8:00 a.m. June 7, 1960, bilirubin content was 13.7 mg. per 100 cc. The patient was less edematous and the intensity of the jaundice had decreased. Her condition rapidly improved over the next few days. On June 13, 1960, the hemoglobin concentration was 10.6 gm. per 100 cc. and the hematocrit 37 per cent. The baby was discharged June 14, 1960, with prescription of Similac® with iron.

When last observed, in April, 1961, the patient had normal physical and mental development.

It was not possible to determine the anti-hr′ titer of the mother in the latter months of pregnancy in order to detect any rise in titer. However, since it was felt that hemolytic disease in this infant was due to hr′ sensitization, serological studies were done. Blood was drawn from the mother, father and four younger children. (The two older sons, ages 16 and 17, were not available.) The mother's anti-hr′ titer of 1:32 gave serological confirmation of the original impression. It is likely that the titer would have been higher if the blood had been drawn in June 1960. Results of the blood studies* are shown in the table below.

							Most Probable Rh Genotype	
		ABO	Rh Factors				Fisher-Race Termi-	Wiener Termi-
Subject	Age	Type	D	C	E	e	nology	nology
Father	40 years	A	+	+	—	+	CDe/cde	R^1r
Mother	37 years	B	+	+	—	—	CDe/CDe	R^1R^1
Sister	12 years	O	+	+	—	+	CDe/cde	R^1r
Brother	7 years	O	+	+	—	—	CDe/CDe	R^1R^1
Sister	3 years	B	+	+	—	—	CDe/CDe	R^1R^1
Patient	3 mo.	O	+	+	—	+	CDe/cde	R^1r

DISCUSSION

The Hr factors are weakly antigenic and consequently only rarely cause antibody formation. However, maternal sensitization to these factors probably occurs more often than is realized. Of the Hr factors, hr′ is the most antigenic and consequently most often stimulates antibody formation. In fact in the Rh-Hr system, hr′ is the most common cause, after Rh_0, of sensitization in pregnancy.[11]

Although it is commonly believed that hemolytic disease due to sensitization to the hr′ factor is mild, it is important to note that many cases of such sensitization reported in the literature were so severe that death resulted. [1-4,6-10,12]

*Done by the Brentwood Laboratories, Los Angeles, California.

The blood transfusions the mother had received earlier may have played some role in sensitizing her to the hr′ factor, although this cannot be substantiated, since Hr typing was not done on the blood she received.

SUMMARY

A case of hemolytic disease of the newborn due to maternal sensitization to the hr′ factor is presented.

Although hemolytic disease of the newborn due to hr′ sensitization is uncommon, such sensitization probably occurs more frequently than is supposed.

Of the Rh-Hr blood factors, hr′ is, after Rh_0, the most common cause of sensitization in pregnancy.

Although some cases of hemolytic disease of the newborn due to the hr′ factor may be mild, other cases may be so severe as to cause death.

Blood transfusions, as well as previous pregnancies, may play a role in sensitizing the mother to the hr′ factor.

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REFERENCES

1. Fisk, R. T., and Brown, A. F.: On the significance of Hr sensitization in Rh antibody determinations, Am. J. Clin. Path., 18:716, 1948.

2. Harris, C.: Erythroblastosis and haemolytic transfusion reactions involving "unusual" blood group factors, Canad. M.A.J., 74:432, 1956.

3. Hopper, H. H.: Transfusion accident and erythroblastosis caused by anti cE, Klin. Wchnschr., 32:975, 1954.

4. Kuhl, I., and Voigt, G. E.: Hemolytic disease due to anti-c (anti hr′-anti I), Zentralbl. Gynak., 76:434, 1954.

5. Levine, P., Burnham, L., Katzin, E. M., and Vogel, P.: The role of iso-immunization in the pathogenesis of erythroblastosis fetalis, Am. J. Obst. & Gynec., 42:925, 1941.

6. Mannherz, K. H., and Krampitz, W.: Fetal erythroblastosis due to anti-c antibody, Zentralbl. Gynak., 79:1329, 1957.

7. Palagi, U.: Hr′ sensitization of Rh-positive mother. Case, Riv. Ist Sieroterap. Ital., 29:270, 1954.

8. Potter, E. L.: Rh . . . Its Relation to Congenital Hemolytic Disease and to Intragroup Transfusion Reactions, The Yearbook Publishers, 1947, pp. 62, 138.

9. Wiener, A. S., and Brancato, G. J.: Erythroblastosis fetalis caused by double sensitization to the factors rh″ and hr′, Anesthesiology, 9:175, 1948.

10. Wiener, A. S., and Brancato, G. J.: Problems in the management of erythroblastosis fetalis with five examples exhibiting unusual serological findings, J. Lab. & Clin. Med., 40:27, 1952.

11. Wiener, A. S., Freda, V. J., Wexler, I. B., and Brancato, G. J.: Pathogenesis of ABO hemolytic disease, Amer. J. Obst. & Gynec., 79:567, 1960.

12. Wiener, A. S., Wexler, I. B., and Brancato, G. J.: Treatment of erythroblastosis fetalis, with special reference to sensitization to Rh subgroup factors other than Rh_0, J. Pediat., 49:381, 1956.